CASE REPORT

Bagcilar Med Bull
DOI: 10.4274/BMB.galenos.2026.44265

Kimura's Disease Presenting as Subcutaneous Masses in Both Elbow Joints: A Case Report and Literature Review

Kimura Hastalığının Her İki Dirsek Ekleminde Subkütan Kitleler ile Ortaya Çıkışı: Olgu Sunumu ve Literatür Derlemesi

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Abstract

Kimura's disease (KD) is a rare chronic inflammatory disorder that can mimic soft tissue tumors or other inflammatory conditions, posing diagnostic challenges. We report a 30-year-old male with a 10-year history of painless subcutaneous masses around both elbows. Laboratory tests revealed marked eosinophilia, and imaging showed bilateral soft tissue swelling with mass formation. Surgical excision was performed, and histopathology confirmed KD, showing lymphoid hyperplasia with eosinophilic infiltration and eosinophilic microabscesses. The patient has remained recurrence-free during the 6-month follow-up. This case highlights the importance of considering KD in the differential diagnosis of long-standing, painless subcutaneous masses to avoid misdiagnosis and unnecessary interventions.

Keywords: Elbow joint, eosinophilia, Kimura's disease

Öz

Kimura hastalığı (KH), yumuşak doku tümörlerini veya diğer enflamatuvar durumları taklit edebilen, tanısal zorluklara yol açan nadir, kronik bir enflamatuvar hastalıktır. Her iki dirsek çevresinde 10 yıllık ağrısız subkütan kitle öyküsü olan 30 yaşında bir erkek hastayı sunmaktayız. Laboratuvar incelemelerinde belirgin eozinofili saptandı ve görüntüleme bulguları bilateral yumuşak doku şişliği ile kitle oluşumunu gösterdi. Cerrahi eksizyon uygulandı ve histopatolojik inceleme, eozinofilik infiltrasyon ve eozinofilik mikroabseler ile birlikte lenfoid hiperplazi göstererek KH tanısını doğruladı. Hasta, 6 aylık takip süresince nüks olmadan izlenmiştir. Bu olgu, uzun süredir mevcut olan ağrısız subkütan kitlelerin ayırıcı tanısında KH'nin dikkate alınmasının, yanlış tanı ve gereksiz girişimlerin önlenmesi açısından önemini vurgulamaktadır.

Anahtar kelimeler: Dirsek eklemi, eozinofili, Kimura hastalığı

Introduction

Kimura's disease (KD) is a rare chronic inflammatory disorder affecting subcutaneous tissues and regional lymph nodes, characterized by eosinophilic infiltration and vascular proliferation (1). Although uncommon, it has been increasingly recognized as a distinct entity, requiring a deeper understanding of its pathophysiology, clinical manifestations, and management strategies.

Case Report

A 30-year-old Asian male presented with a 10-year history of painless, movable subcutaneous masses in both elbows. The lesions gradually increased in size, and the patient had not sought treatment. He denied any other medical history. Examination revealed localized swelling on the medial aspects of both elbows with mild erythema, elevated local temperature, minimal tenderness, and full joint mobility.

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Received: 20.01.2026 **Accepted:** 27.04.2026 **Epub:** 30.04.2026

Cite this article as: Yang W, He C. Kimura's disease presenting as subcutaneous masses in both elbow joints: a case report and literature review. Bagcilar Med Bull. [Epub Ahead of Print]

Laboratory testing demonstrated marked eosinophilia (33.1%, reference 0.0-5.0%; absolute count 2.50×10^9/L, reference $0.00\text{-}0.50\times10^9$/L), while other leukocyte subsets, uric acid, coagulation parameters, tumor markers, and erythrocyte sedimentation rate were within normal limits. Axial non-contrast computed tomography of the elbow joints demonstrated bilateral subcutaneous masses, measuring approximately 32×23 mm on the right and 34×23 mm on the left, with slightly indistinct margins, homogeneous density, and mild surrounding soft-tissue swelling (Figure 1A, B). Magnetic resonance imaging (MRI) of the elbows showed that the lesions demonstrated high signal intensity on diffusion-weighted imaging (DWI) (Figure 1C), with corresponding low signal on the apparent diffusion coefficient map (Figure 1D). The lesions exhibited homogeneous high signal intensity on T2-weighted imaging (T2WI) (Figure 1E, F) and homogeneous low signal intensity on T1-weighted imaging (T1WI) (Figure 1G, H). The masses were well circumscribed without invasion of adjacent muscles. The patient underwent surgical excision of the lesions. Histopathology revealed lymphoid hyperplasia with prominent eosinophilic infiltration, proliferation of endothelial venules in the interfollicular regions, and eosinophilic microabscess formation, confirming KD (Figure 2A, B). Postoperatively, the patient has remained recurrence-free during regular 6-month follow-up. Informed consent was obtained from the patient for the anonymous use and publication of clinical and imaging data.

Discussion

KD, or eosinophilic hyperplastic lymphogranuloma, is a rare benign granulomatous disorder originating in the dermis, subcutaneous tissue, and lymph nodes (2). It most commonly affects young Asian males and typically presents as painless subcutaneous nodules in the head and neck, often accompanied by lymphadenopathy. Peripheral eosinophilia is common, highlighting an immunological component in its pathogenesis. Although the etiology remains unclear, immune dysregulation is thought to play a pivotal role, with eosinophilic infiltration and cytokine-driven vascular proliferation as key features (2,3).

The prolonged presence of subcutaneous masses over a decade in atypical locations such as the elbows is unusual; only a few cases have been reported (4,5).

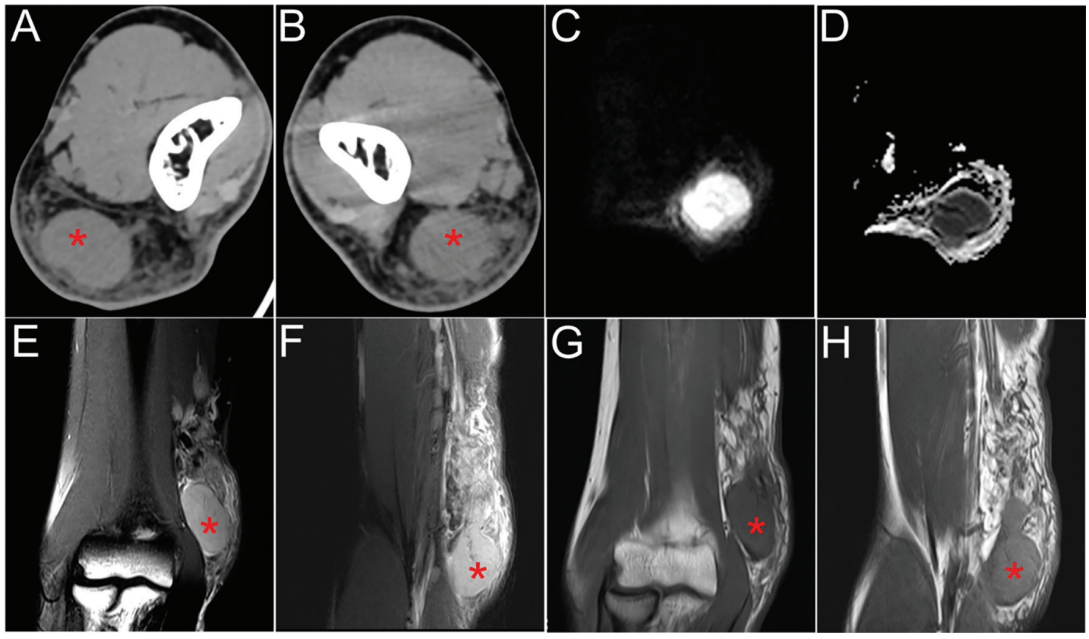

Figure 1. Axial non-contrast CT of the elbow joints demonstrated subcutaneous soft-tissue masses around the left elbow (A, red asterisk) and right elbow (B, red asterisk), with slightly indistinct margins, homogeneous density, and mild surrounding soft-tissue swelling. On DWI, the lesions showed high signal intensity (C), with corresponding low signal on the ADC map (D). Coronal T2WI fat-suppressed images revealed slightly hyperintense subcutaneous masses at the left elbow (E, red asterisk) and right elbow (F, red asterisk). Coronal T1WI demonstrated isointense subcutaneous masses at the left elbow (G, red asterisk) and right elbow (H, red asterisk), with clear delineation from adjacent muscles and mildly obscured surrounding fat planes

ADC: Apparent diffusion coefficient, DWI: Diffusion-weighted imaging, CT: Computed tomography, T1WI: T1-weighted imaging, T2WI: T2-weighted imaging

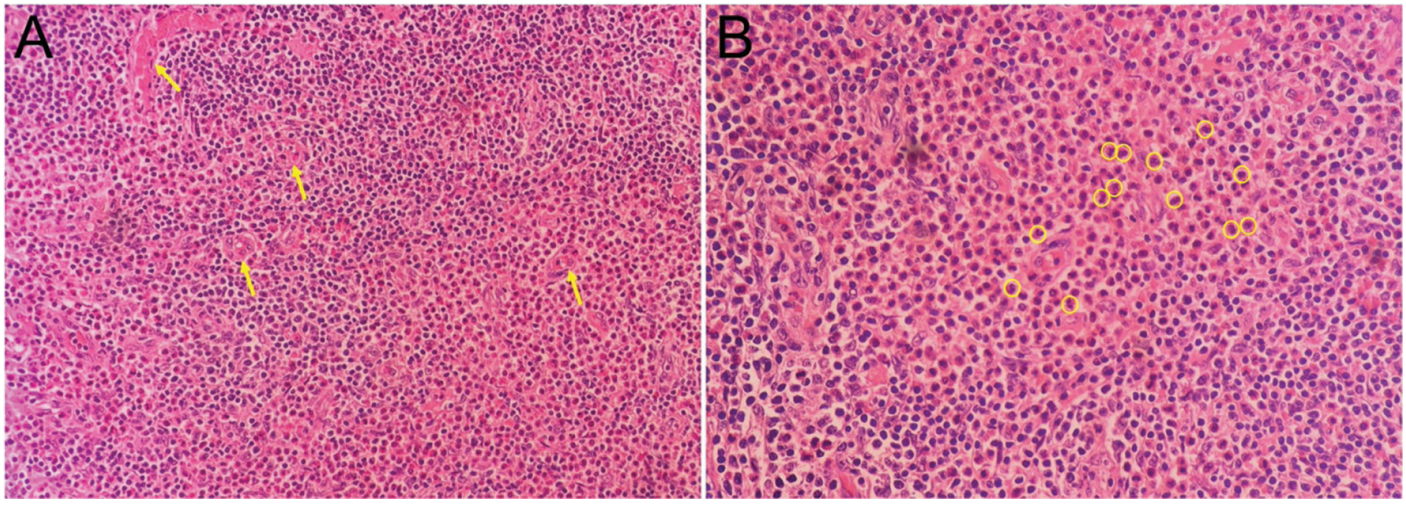

Figure 2. At low magnification (A, H&E ×200), lymphoid hyperplasia with prominent eosinophilic infiltration is observed. Proliferation of small venules is evident in the interfollicular areas (yellow arrows), along with the formation of eosinophilic microabscesses. At high magnification (B, H&E ×400), numerous eosinophils (yellow circles) are present, exhibiting bilobed nuclei and cytoplasm densely packed with coarse eosinophilic granules

H&E: Hematoxylin and eosin

Such atypical presentations expand the recognized spectrum of KD and underscore the importance of considering it in the differential diagnosis of chronic subcutaneous masses outside the head and neck region. The imaging manifestations are enlarged lymph nodes with uniform density, no necrosis, uniform enhancement, and the presence of a lymph node hilum, which can be differentiated from malignant lymph nodes (6). Xie et al. (7) reported five cases of KD occurring at uncommon sites, four of which involved the upper arm. The imaging findings demonstrated homogeneous masses; MRI showed isointense signal on T1WI and hyperintense signal on T2WI. Although typically reported as isointense on T1WI, slight hypointensity may also be observed, as in our case. Some lesions exhibited mild diffusion restriction on DWI. Contrast-enhanced scans revealed mild to marked enhancement. Varying degrees of perilesional swelling were observed, while adjacent muscles, bones, and joints were not involved. Our case is consistent with these previous reports. Histopathological findings in KD typically include lymphoid hyperplasia with eosinophilic infiltration. In this case, high endothelial venule proliferation and eosinophilic microabscesses further supported the diagnosis.

Management of KD is multidisciplinary. Corticosteroids remain the first-line therapy, effectively reducing eosinophilic infiltration and symptom burden (8). Surgical excision is indicated for localized, symptomatic lesions. Immunomodulators may be considered for recurrent or refractory cases; anti-interleukin (IL)-5 antibody and IL-4 receptor α antibody have been successfully used (9-11). Although KD is benign, relapse can occur, highlighting the importance of long-term follow-up. Prognosis is generally favorable with timely and appropriate intervention.

Conclusion

This case highlights an atypical presentation of KD with subcutaneous masses in the elbows. Combining clinical, imaging, and histopathological findings allowed for accurate diagnosis and effective management. Recognizing such atypical manifestations improves diagnostic accuracy, avoids unnecessary interventions, and informs tailored treatment strategies for this rare disease.

Ethics

Informed Consent: Informed consent was obtained from the patient for the anonymous use and publication of clinical and imaging data.

Footnotes

Authorship Contributions

Concept: W.Y., C.H., Design: W.Y., Data Collection or Processing: W.Y., Analysis or Interpretation: C.H., Literature Search: W.Y., C.H., Writing: W.Y.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

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